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Calcifying Epithelial Odontogenic Cyst: An Uncommon Variant

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ABSTRACT: The Calcifying Epithelial Odontogenic Cyst (CEOC) is a developmental odontogenic cyst, which was first categorized as a distinct entity by Gorlin in 1962. It is an unusual and unique lesion, which shows the characteristics of both a solid neoplasm and a cyst. We present a case report of calcifying epithelial odontogenic cyst in a 69-year-old male who presented with a painless unilateral swelling of jaw. The lesion was surgically removed and histopathological examination revealed a tumor with predominance of ghost cells and some amount of dentinoid tissue.

Keywords: - Calcifying cystic odontogenic tumor, Calcifying epithelial odontogenic cyst, Ghost cells.

I. INTRODUCTION

Calcifying epithelial odontogenic cyst (CEOC) is an uncommon benign odontogenic lesion, which represents about 2% of all odontogenic tumors and cysts. It was first categorized as distinct entity by Gorlin in 1962.1They initially considered it as the oral analogue of the cutaneous calcifying epithelioma of Malherbe, but later labeled it as CEOC1. It is an unusual and unique lesion which occupies a spectrum ranging from a cyst to an odontogenic tumor, with characteristics of a cyst alone or those of a solid neoplasm2. In 1992, the World Health Organization classified CEOC within the groups of neoplasms and tumors that originate from odontogenic tissues, but confirmed that most of the cases are non-neoplastic and has a potential for continuous growth.3 A lot of confusion and disagreement is present in the terminology and classification of CEOC. Some investigators have considered CEOC as a tumor with a tendency for marked cyst formation in which the concept, called "monistic" by Toida has led some researchers to substitute the terms "calcifying ghost cell odontogenic tumor" or "cystic calcifying odontogenic tumor" for that of CEOC. In addition, a "dualistic" approach has been suggested that CEOC can contain two entities: 4 1. Cyst: calcifying ghost cell odontogenic cyst 2. Neoplasm (benign: Calcifying ghost cell odontogenic tumor; or malignant: Malignant calcifying ghost cell odontogenic tumor). CEOC most often occurs as a central(intra-osseous) lesion, whereas peripheral (extra-osseous) in the soft tissue is rare.8 It usually appears as painless, slow growing tumor affecting equally the maxilla and mandible, with predilection for the anterior segment (incisor/canine area). It generally affects young adults in the third to fourth decades without gender predilection. Radio graphically CEOC is usually a mixed lesion, appears either as a unilocular or multilocular well-defined radiolucency with irregular calcified bodies of varying sizes. Within it and may be associated with an odontome or an unerupted tooth.5 The most characteristic histopathological feature is the presence of ghost cells with the propensity to calcify.5 The present paper reports a case of CEOC in a 69-year old male with brief review of relevant literature.

II. CASE REPORT

A 69 year old male patient reported to the department of oral medicine and radiology with the chief complaint of swelling in the lower right back tooth region of jaw since 2 years. Patient reported that swelling started 2 years back and gradually increased to attain the present size. No history of pain or other associated signs and symptoms were present. His past medical and dental history were non-contributory. General examination revealed a moderately built and nourished individual with steady gait and vital signs within normal range.

Extraorally no apparent facial asymmetry was noticed. Intraoral examination revealed a diffuse swelling present in the right lower edentulous alveolar area measuring 1.5×2cm in diameter, extending anteriorly up to gingivobuccal sulcus of missing 45,posteriorlyto the retromolar trigone and laterally causing obliteration of buccal vestibule, with no pathology noticed on the overlying mucosa. The swelling was bony hard in consistency with a windowing in the anterior aspect and surface was smooth and non-tender on palpation (Figure 1).

No tenderness or mobility of adjacent teeth was noted. Aspiration yielded a brownish yellow colored fluid (Figure 2).

On the basis of history and clinical examination, provisional diagnosis of benign odontogenic tumor of jaw was given. Various differential diagnosis of keratocystic odontogenic tumor, calcifying epithelial odontogenic tumor, ameloblastoma, dentigerous cyst were included.

Further the patient was subjected to radiographic examinations. Intraoral periapical radiograph revealed a unilocular radiolucency with a well-defined sclerotic border with superior cortical border elevation. The internal structure appeared mixed radiopaque-radiolucent. Mandibular cross-sectional occlusal radiograph revealed bicortical plate expansion with intact cortical boundary.

Panoramic radiography revealed a well-defined radiolucency involving right body and ramus of the mandible extending from distal aspect of missing 45 to the junction of body and ramus of mandible, measuring 4×2cm with sclerotic border in anterior aspect and poorly differentiated border in the posterior aspect with erosion of superior border (Figure 3). The internal structure appeared granular with inferiorly displaced inferior alveolar canal. To know the clear extension of the lesion, cone beam computed tomography was performed which revealed a well-defined predominantly hypodense lesion with localized hyperdense areas (Figure 4a, b, c).

The lesion was treated conservatively with careful enucleation and curettage under LA, followed by application of carnoy's solution. Histopathological report showed a lining of epithelium resembling odontogenic apparatusand numerous ghost cells. Basal cells resembled ameloblasts and suprabasal cells resembled stellate reticulum. Some of ghost cells showed dystrophic calcifications. Irregular foci of tissue resembling dentine, which were atubular were observed in connective tissue (Figure 5 a, b). On the basis of histopathological report, final diagnosis of CEOC was given.

Post-operative healing was uneventful. The patient was kept under regular follow-up. One year post-operative evaluation was done and no evidence of reccurence was seen. Radiographs was taken and which showed presence of healthy bone formation (Figure 6).

III. DISCUSSION

The CEOC is an uncommon odontogenic lesion that demonstrates considerable histologic diversity and presents variable clinical behavior. Although it is widely considered as a cyst, some investigators prefer to classify it as a neoplasm.6

Calcifying epithelial odontogenic cyst's are thought to be developed from the reduced enamel epithelium or remnants of odontogenic epithelium in the follicle, gingival tissue or bone.1 The epithelial lining of a CEOC appears to have the ability to induce the formation of dental tissues in the adjacent connective tissue wall, and thus may be associated with other odontogenic tumors.7

The age of these patients may range from 5 to 92 years, with a peak incidence in the second decade of life.8 Literature state a bimodal age distribution, with a second peak in the sixth-seventh decade of life.2The lesion has no sex predilection and is equally distributed between the maxilla and mandible. It commonly occurs anterior to first molar region in which 75% of cases are in incisor-canine region or inter-canine region, where the cyst sometimes manifests as a pericoronal radiolucency.2 Clinically, the lesion usually presents as an asymptomatic swelling causing a bony hard expansion of the jaw. In the present case, the age of occurrence is consistent with that described in literature but not in accordance with site. Radiographically, the lesion present in an unilocular form with well-defined corticated margins but 5-13% of cases are multilocular.8 The internal structure may vary in appearance – it may be completely radiolucent, but is usually mixed (radiolucent-radiopaque), and may have scattered irregular sized calcifications producing a variable range of opacities (salt and pepper type of pattern), or may even show large solid amorphous masses.2 Central CEOC has been reported to be associated with an odontoma in 24-35% of cases and with impacted teeth in 35% cases, usually a canine. Unilocular lesions may mimic dentigerous, radicular or residual cysts while multilocular lesions may resemble ameloblastomas or odontogenic keratocysts.7Radiographically the present case shows a unilocular radioluceny with some radiopaque foci in the anterior aspect. Expansion of the buccal and lingual cortical plates was evident.

The histological features of a classical CEOC include a cystic cavity with a fibrous capsule, with a lining of odontogenic epithelium that is 6-8 cells thick. The basal layer is made up of ameloblast-like columnar or cuboidal cells with hyperchromatic nuclei polarized away from the basement membrane. The overlying loosely arranged epithelial layer shows similarity to the stellate reticulum of the enamel organ.2 The typical microscopic characteristic of this lesion is the presence of variable amounts of aberrant epithelial cells, devoid of nuclei, which are eosinophilic and retain their basic cell outline, and are called "ghost cells."3These ghost cells may undergo calcification and lose their cellular outline to form sheet-like areas of calcified keratin. The nature of ghost cell change may be due to the effect of coagulative necrosis and dystrophic calcification or may be a form of normal or abnormal keratinization of the odontogenic epithelium. Ghost cells are also seen in

odontoma, ameloblastoma, craniopharyngioma, and other odontogenic tumors.2 Present case showed similar microscopic findings with presence of cystic epithelial lining with ameloblast-like cells, ghost cells, dystrophic calcification and irregular areas of dysplastic dentine. The CEOC is treated conservatively by surgical enucleation and recurrences are very uncommon, as recurrence depends on the completeness of cyst removal. The malignant transformation of a preexisting benign CEOC can occur but is extremely uncommon.3In the present case, surgical enucleation of the lesion was done and patient was kept under regular follow up.

IV. FIGURES AND LEGENDS

Figure 1: Intraoral photograph showing swelling in right lower edentulous alveolar ridge.

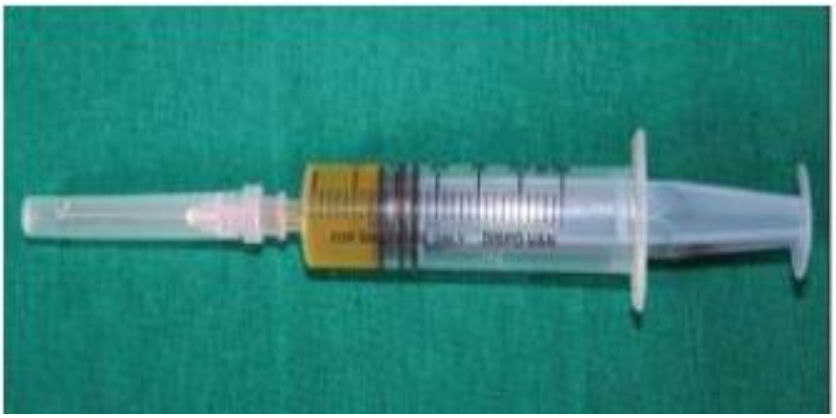

Figure 2: Brownish yellow colored fluid obtained on aspiration

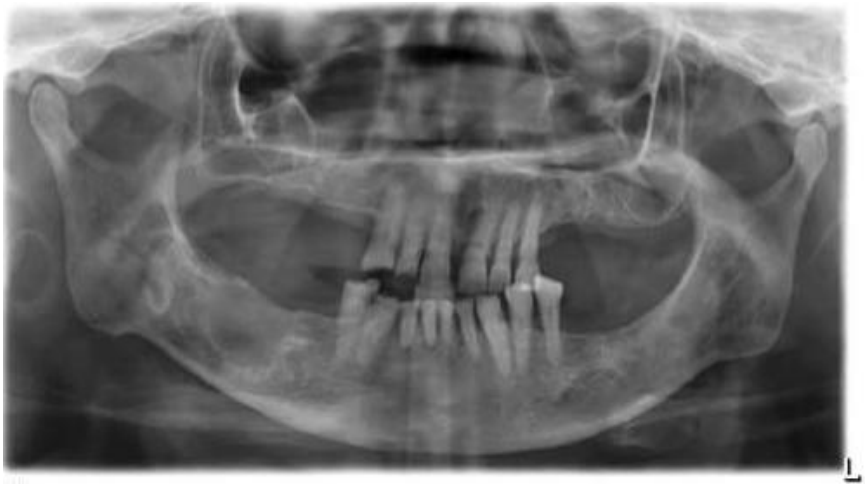

Figure 3: Panoramic radiography showing radiolucency in the right lower edentulous alveolar ridge extending from distal aspect of missing 45 posteriorly to the junction of ramus and body of mandible with flecks of radiopacities in the anterior aspect.

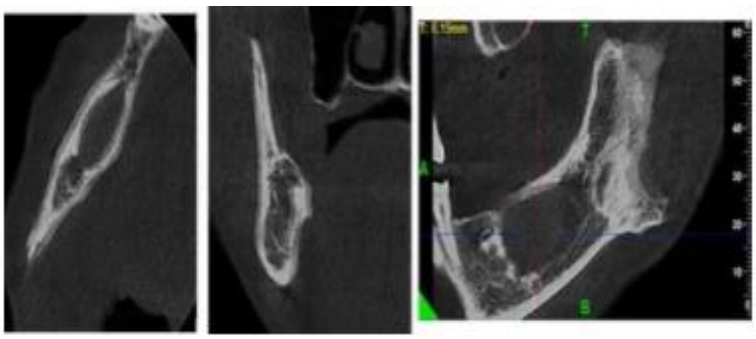

Figure 4a, 4b, 4c: CBCT images showing well-defined predominantly hypodense lesion with localized hyperdense areas in axial, coronal and sagittal sections respectively.

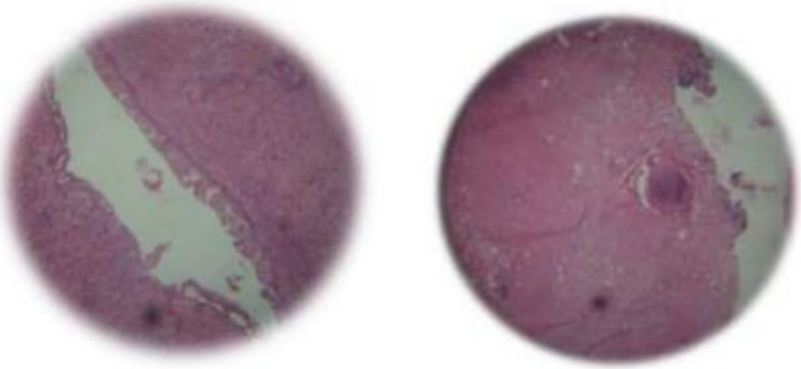

Figure 5a, 5b: Photomicrograph showing basal layer of columnar cells with hyperchromatic nuclei and presence of ghost cells with dystrophic calcifications respectively.

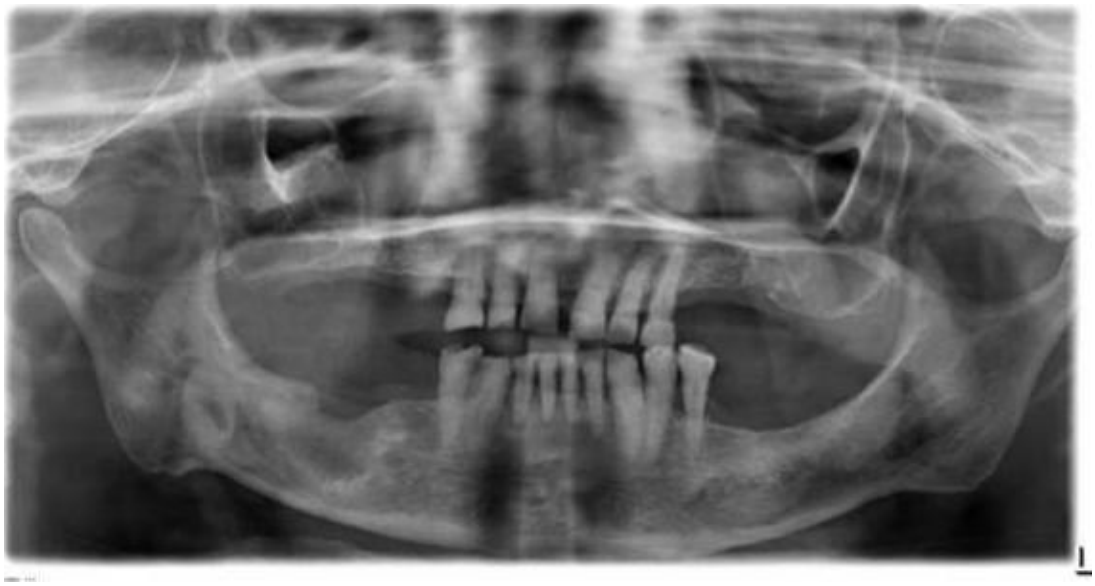

Figure 6: One year postoperative panoramic radiography showing new healthy bone formation in the surgical site.

V. CONCLUSION

Calcifying epithelial odontogenic cyst may mimic various odontogenic and non odontogenic lesions, making diagnosis very difficult and definitive diagnosis can only be made histologically. Therefore, careful understanding of specific clinical and histological features is crucial. Long term follow-up visits are recommended especially if adjacent structures are also affected.

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